Rare Cause of Severe Dyspnea After Tracheotomy-Negative Pressure Pulmonary Edema

Case Report

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Abstract

Deep neck infections are serious conditions and can present with acute upper airway obstruction. Our priority in the treatment is to ensure airway safety, and tracheotomy may be needed to overcome the upper airway obstruction. Unceasing dyspnea after tracheotomy should suggest serious pulmonary pathologies in patients with upper airway obstruction due to deep neck infection. Acute/chronic obstruction resolved after tracheotomy or upper respiratory tract surgical procedures of obstructive sleep apnea patients can turn into severe dyspnea with pulmonary edema. In this report, we present a 46-year-old male patient with negative pressure pulmonary edema as a complication of tracheotomy. The tracheotomy was performed due to severe upper airway obstruction secondary to a deep neck infection. The importance of early diagnosis and prompt treatment of this rare entity after unceasing dyspnea despite tracheotomy is discussed in the light of the current literature

Keywords: Obesity, pulmonary edema, pulmonary gas exchange, sleep apnea syndrome, tracheotomy, case report

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Introduction

In deep neck infections (DNIs), the first priority of treatment is to ensure airway safety, and then it is necessary to review options such as antibiotic therapy and surgical drainage (1). Urgent and/or elective tracheotomy requirement varies between 2.7 and 32.9% in patients experiencing upper airway obstruction after DNIs (1, 2).

Negative pressure pulmonary edema (NPPE) is an important pulmonary complication that is rarely seen after tracheotomy. Upper respiratory tract operations, obesity, short neck, and obstructive sleep apnea syndrome (OSAS) are reported among the factors that increase the risk of developing NPPE (3, 4).

Ensuring airway safety is the first priority in DNI patients and tracheotomy is one of the treatment options (5). In this case report, we present a case of NPPE, a rare postoperative complication that could be mortal if misled. Written and verbal consent was obtained from the patient for this report.

Case Presentation

A 46-year-old male patient presented to our clinic with worsening tonsillopharyngitis while on antibiotics

for five days. Physical examination and computed tomography (CT) revealed multiple abscess foci in the deep neck spaces and severe trismus (Figures 1, 2). Preoperative thoracic CT imaging was within normal limits. The patient had diabetes mellitus, OSAS (with the use of continuous positive airway pressure device), heart rhythm disorder, and obesity (body mass index: 37). The patient was taken to the operating room for abscess drainage and tracheotomy. The preoperative risk was stated by the anesthesiologists as American Society of Anesthesiologist 3E. The anesthesiologist predicted the need for an intensive care unit for probable postoperative pulmonary complications due to chronic airway obstruction with OSAS and additional acute respiratory distress with upper airway edema.

Since the patient had trismus, the preoperative examination was performed together with the anesthesiologist, and it was thought that sudden collapse might occur after the administration of muscle relaxants. Therefore, upon the recommendation of the anesthesiologist, the patient was not intubated but a tracheotomy was performed with local anesthesia.

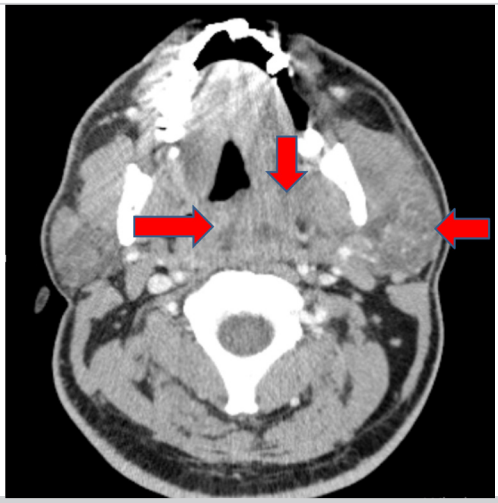

Figure 1. Neck CT image showing peritonsillar-parapharyngeal-intraparotidal multifocal abscess areas (arrows)
CT: Computed tomography

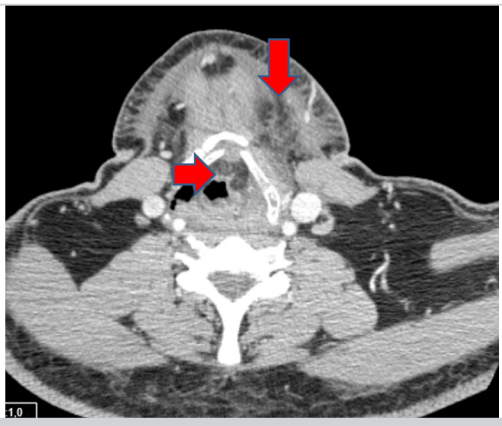

Figure 2. Neck CT, axial section showing abscess foci extending to the larynx (arrows)
CT: Computed tomography

The general anesthesia procedure was then initiated by ventilation through the tracheotomy tube. Emergency tracheotomy and multiple (peritonsillar, parapharyngeal, submandibular, intraparotidal, and intraoral retropharyngeal) abscess drainage were performed, and pathology and abscess culture samples were taken under general anesthesia. Although the patient had an uncomplicated tracheotomy, he had severe dyspnea and low oxygen saturation (80%) after arising from general anesthesia. He was taken to the intensive care unit with the recommendation of the anesthesiologist. Sputum containing very dense foam was aspirated from the tracheotomy cannula and oxygen support of 10 lt/min was administered through the tracheostomy cannula. Thoracic CT imaging revealed predominantly centrally located multifocal ground glass areas and interlobular septal thickenings in both lung parenchyma without mediastinitis findings (Figure 3). Coronavirus disease-2019 was ruled out and differential diagnoses of alveolar hemorrhage and NPPE were considered.

As the patient did not benefit from the oxygen administered via the cannula, decision was made to apply invasive mechanical ventilator (IMV) through the tracheostomy cannula, furosemide infusion for diuresis, and empirical meropenem treatment were started. A control CT was taken 48 hours after the patient's oxygen saturation reached 90%, and it was determined that the abscess foci in the parapharyngeal space of the neck continued and the ground glass appearance of the lung regressed but did not completely disappear. The patient was once more taken to the operating room for abscess drainage. Postoperatively, he was connected to an IMV with the recommendation of the pulmonology department. After three days of IMV, the patient's oxygen saturation improved and his pulmonary symptoms regressed.

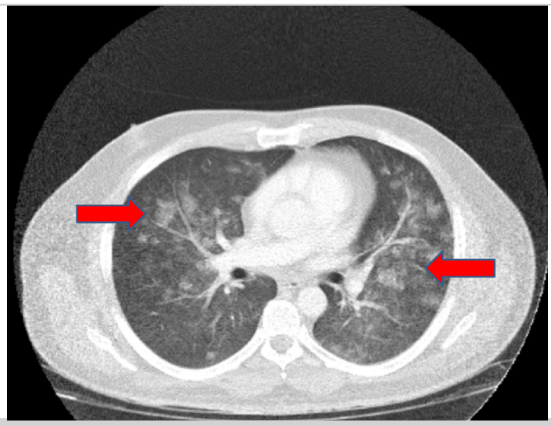

Figure 3. NPPE thoracic CT axial image (arrows)
NPPE: Negative pressure pulmonary edema, CT: Computed tomography

Abscess drainage was performed for the third time 10 days after the second surgery when the patient was suspected of collection in the retropharyngeal area due to fluctuating C-reactive protein and neutrophil levels. The patient was discharged after 30 days with a decannulated tracheotomy after his laryngeal edema regressed and pulmonary problems were completely relieved. No problems were observed in the follow-up visits.

Discussion

The main mechanism of NPPE is that acute closure of the airway causes negative intrathoracic pressure increase caused by forced inspiration against the upper airway, increased pulmonary capillary hydrostatic pressure, and hydrostatic pulmonary edema. Two distinct clinical mechanisms can be identified for NPPE. Type I NPPE occurs soon after the onset of the acute upper airway obstructive process. Type II NPPE develops after the resolution of chronic upper airway obstruction. Most cases of NPPE described in the literature are type I NPPE that develop after the onset of an acute episode of upper airway obstruction. It relies on a different mechanism and process in type II NPPE. Relief of chronic airway obstruction caused by various conditions such as adenotonsillar hypertrophy, laryngotracheal neoplasm, and thyroid goiter can result in type II NPPE (6, 7).

In the presence of forced inspiration, negative pleural pressure can rise well above the normal value. The result is increased venous return to the right heart and dilation of the right ventricle. The interventricular septum shifts to the left, and cardiac output decreases as a result of diastolic dysfunction. Fluid accumulation in the alveoli and pulmonary edema are observed as a result of impaired microvascular circulation and increased pulmonary capillary permeability (8). This often occurs immediately after upper airway obstruction, but symptoms can take up to six hours to appear (3). Pulmonary infiltrates and interstitial pulmonary edema can be seen on chest X-ray or CT. The presence of clinical and radiological findings suggestive of pulmonary edema and the absence of cardiac pathology should suggest the diagnosis of NPPE.

Aspiration pneumonia, cardiogenic pulmonary edema, and mediastinitis are important in its differential diagnosis (9). Type II NPPE is based on a mechanism of chronic upper airway obstruction. Relief of chronic upper airway obstruction caused by various conditions such as adenotonsillar hypertrophy can result in type II NPPE (7). For OSAS patients, respiratory track surgical procedures can also result in pulmonary edema in a similar manner. Our patient had risk factors such as obesity, short neck, upper airway obstruction, tracheotomy, and OSAS. Foamy sputum, low oxygen saturation, severe acute dyspnea, acute severe pulmonary infiltrates, and edema on CT enabled us to make the diagnosis. One of the most common pulmonary pathologies that cause dyspnea in DNI patients is mediastinitis. However, the differential diagnosis was made with thoracic CT in our patient, and mediastinitis was ruled out. The main goal in the treatment of NPPE is to ensure airway patency and adequate oxygenation. Oxygen saturation should be maintained above 90%. In mild cases, an oxygen mask was found sufficient. However, if the patient's clinical and oxygen values do not improve, noninvasive mechanical ventilation (continuous positive airway pressure or bilevel positive airway pressure) should be applied (9). Restriction of intravenous crystalloids, maintenance of normal intravascular volume and serum oncotic pressure with colloids, and use of diuretics may be recommended in the treatment of NPPE. Some authorities recommend steroids in the NPPE treatment regimen. However, the use of steroids for treatment still seems controversial (7).

Conclusion

NPPE can develop in DNI cases after upper airway obstruction or after tracheotomy for the treatment of upper airway obstruction. We share this rare complication of tracheotomy to contribute to the literature.

Informed Consent: Written and verbal consent was obtained from the patient for this report.

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Authorship Contributions

Surgical and Medical Practices: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö., Concept: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö., Design: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö., Data Collection and/or Processing: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö., Analysis and/or Interpretation: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö., Literature Search: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö., Writing: K.K.B., O.B., C.G.C.E., H.G., O.İ., E.S.Ö.

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Main Points

- Deep neck infections may present with severe upper airway obstruction.
- The primary priority of management is to secure the airway.
- Sudden removal of airway obstruction in patients with risk factors can cause negative pressure pulmonary edema.
- Negative pressure pulmonary edema is a mortal condition and should be treated quickly with a multidisciplinary approach.

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